Case Report

Sinonasal Teratocarcinosarcoma: A Rare Case Report

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Abstract

Background: Teratocarcinosarcoma is a type of malignancy that typically arises from germ cells. A teratocarcinoma in the nose would present as an unusual or rare clinical case, requiring imaging and biopsy for confirmation. It constitutes less than 1% of all cancers and approximately 3% of all malignancies of the head and neck region; only a few hundred cases have been reported worldwide. Because of its rarity, clinical presentation, and treatment challenges, it is essential to report and study cases of nasal teratocarcinoma.

Case Presentation: We here report a case of Sinonasal Teratocarcinosarcoma (SNTCS), a 55-year-old female with a complaint of right-sided nasal blockage for over a month with a nasal bleed for one day. On local examination, there was swelling of the right alar region and dorsum with DNS to the left side and tenderness in all paranasal sinuses. There was no fogging on the right side. On anterior rhinoscopy, a chocolate-brown polypoidal mass obscuring the view of the floor, roof, cavity, and middle turbinate was seen. On histopathological examination, it was diagnosed as teratocarcinosarcoma of the nasal cavity. Immunohistochemistry confirmed the diagnosis. On follow-up, her condition deteriorated and she succumbed to death in the next 6 months.

Conclusion: Teratocarcinosarcoma is a rare and aggressive sinonasal tumor with a hypothesized origin from somatic pluripotent stem cells of the nasal cavity. Its early diagnosis is important as it carries a poor prognosis.

Keywords: Teratocarcinosarcoma; Nasal cavity; Polyp; SNTCS

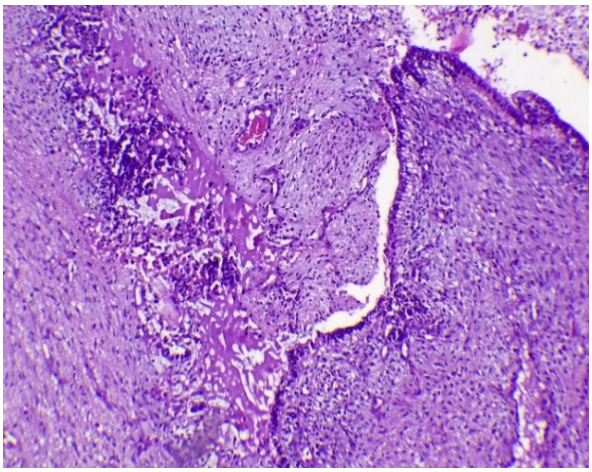
Introduction

Sinonasal teratocarcinosarcoma (SNTCS) is a unique, rare, and aggressive malignancy of the anterior skull base first reported by Heffner and Hyams in 1984 [1, 4]. Typically arising in paranasal or ethmoid sinuses, this neoplasm poses substantial diagnostic challenges given its heterogeneous composition [2]. It exhibits a highly malignant nature and poor prognosis [3].

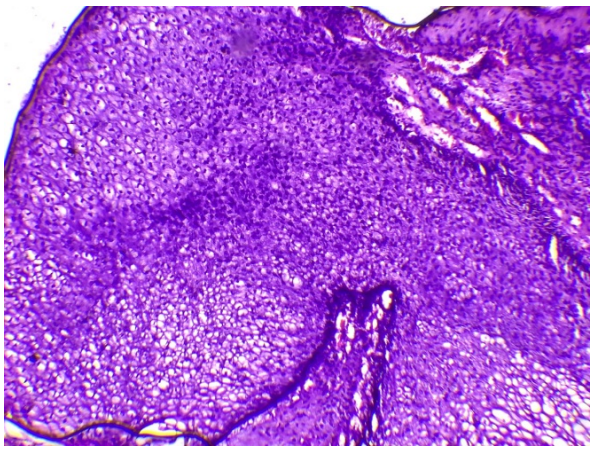
Clinical examination of tumors includes: (a) Nasal obstruction and hyposmia resulting from local tumor growth; (b) Epistaxis, fetid odor in the nasal cavity, and increased nasal secretions due to rapid tumor growth leading to necrosis; and (c) Invasion of adjacent tissues by the tumor can result in facial edema, lacrimation, proptosis, visual impairment, headaches, and alterations in consciousness and behavior. Physical examination may reveal a nasal mass with an ill-defined morphology that cannot be distinguished from polyps or papillomas. The disease typically follows a relatively short course, lasting 2-10 weeks but occasionally extending up to 2 years [14]. It is a histologically complex tumor, comprised of teratoma and carcinosarcoma of varying maturity without malignant germ cell components such as embryonic carcinoma, choriocarcinoma, or seminoma [5, 7]. SNTCS tends to occur more frequently in adult males than in females (7:1) [6]. This case report aims to present a rare and aggressive case of sinonasal teratocarcinosarcoma (SNTCS), highlighting its diagnostic challenges, histopathological features, and multidisciplinary approach to treatment. Given the tumor's rarity and poor prognosis, we seek to contribute to the existing literature by discussing clinical presentation, imaging findings, treatment strategies, and follow-up details, thus enhancing awareness and guiding future therapeutic approaches for this malignancy.

Case Report

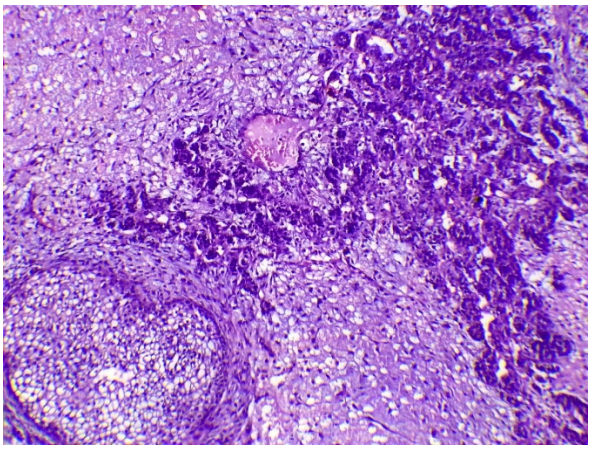
A 55-year-old female complained of a right-sided nasal blockage for one month and a nasal bleed for one day. On local examination, there was swelling of the right alar region and dorsum, DNS to the left side, and tenderness in all paranasal sinuses. There was no fogging on the right side. On anterior rhinoscopy, a chocolate-brown polypoidal mass was obscuring the view of the floor, roof, cavity, and middle turbinate. On computed tomography scan-PNS revealed polyposis involving all the paranasal sinuses with nasoethmoidal and nasomaxillary extension, deviation of the nasal septum towards the left with bilateral inferior turbinate thickening. The clinical diagnosis rendered was telangiectatic polyp. An excisional biopsy was done and sent for histopathological examination. On histopathological examination, it was diagnosed as teratocarcinosarcoma of the nasal cavity. Immunohistochemistry confirmed the diagnosis. It showed Vimentin (strong, diffuse), SALL4, CK (strong, diffuse), and NSE (strong, diffuse) positivity.



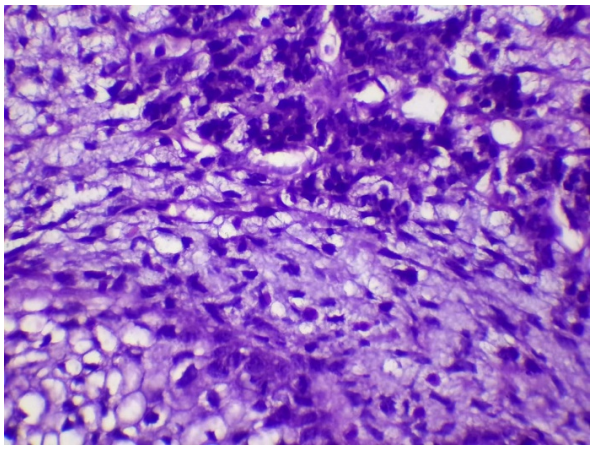
(a) Lining of sinonasal epithelium (100X).



(b) Fetal squamous epithelium (100X).



(c) Epithelial and mesenchymal component (100X).

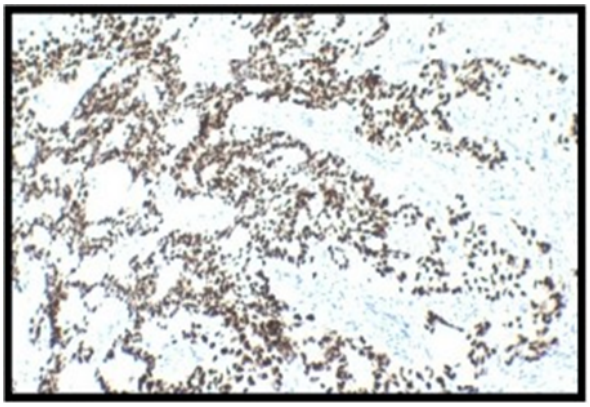


(d) Epithelial component in the form of glandular arrangement (400X).

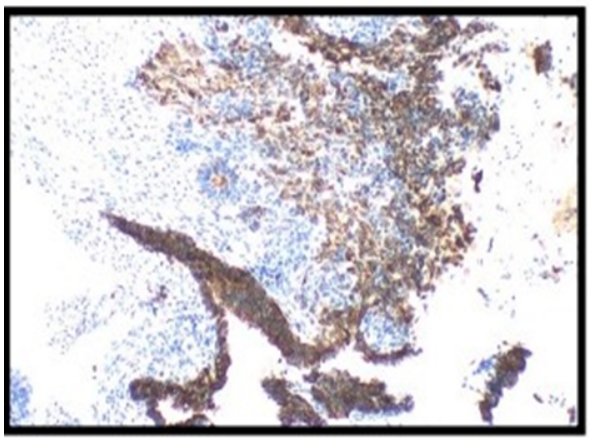
Figure 1: Microscopic examination findings.

Discussion

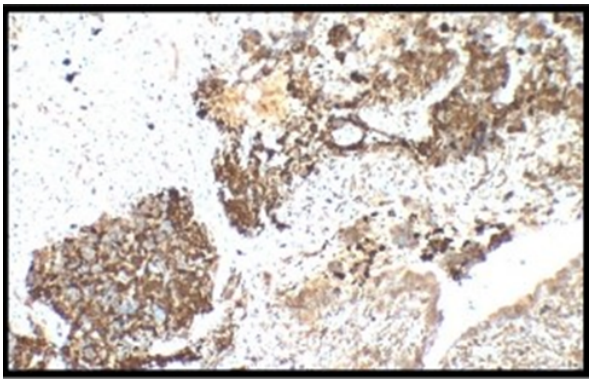
Teratocarcinosarcoma is a rare and aggressive sinonasal tumor with a hypothesized origin from somatic pluripotent stem cells of the nasal cavity. Only 127 cases have been reported to date [8]. It was first described in 1983 by Shanmugaratnam as teratoid carcinosarcoma [15]. Heffner and Hyams were the first to coin the term “teratocarcinosarcoma” based on the clinicopathological study of 20 cases of sinonasal tract neoplasms [15]. It is almost exclusively seen in adults, with only three pediatric cases being reported [15]. Because of the infrequency and the complex phenotypic elements, these lesions are often misdiagnosed, leading to management difficulties. SNTCS is an exceptionally rare entity occurring in the 5th to 6th decade of life with male predilection and less than two years median survival. Other sites of teratocarcinosarcomas are the testis, ovaries, and uterine cervix. Local recurrence with intracranial extension is relatively common, while rare distant metastasis eventually accentuates morbidity and mortality. The aggressiveness of the tumor increases after each recurrence. Hence, this underscores the importance of early diagnosis with a prompt multimodality treatment approach, i.e., surgery followed by



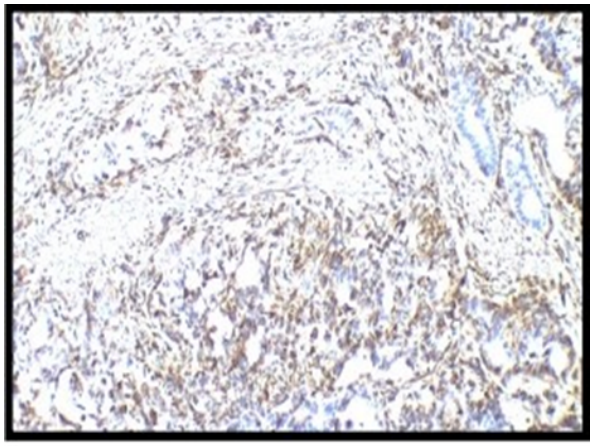
(a) Vimentin positive.



(b) CK positive.



(c) SALL4 positive.



(d) NSE positive.

Figure 2: Immunohistochemistry positive markers.

adjuvant chemoradiation therapy along with diligent follow-up. Extensive sampling is of pertinence to avoid an erroneous diagnosis of other sinonasal tumors. Early diagnosis and management before regional spread can give a better prognosis. This case was followed up and was found to have two recurrences within 5 months without any post-operative chemotherapy and radiotherapy, and after every recurrence, there was a deterioration in the general health of the patient. Clinically, these patients generally present with epistaxis or nasal obstruction. Sinonasal teratocarcinosarcoma microscopically consists of primitive neuroepithelial elements with various malignant epithelial and mesenchymal components, probably originating from stem or pluripotent progenitor cells with multidirectional differentiation [9]. Usually, squamous cell or adenocarcinoma is seen, and the mesenchymal component shows spindle, smooth, skeletal muscle, cartilage, and bone features.

It was previously reported that immunohistochemical results are helpful for the diagnosis of SNTCS. These included positive staining for epithelial markers (cytokeratin and EMA), the undifferentiated shuttle sarcoma/smooth muscle marker vimentin, undifferentiated or primary tumor component markers (CD99 and NSE), and negative staining for smooth muscle actin and desmin. These staining patterns are consistent with the immunohistochemical findings in our case [10]. Smith et al. reported a study of 10 cases where all were males ranging from 39 to 65 years. All presented with a brief history of epistaxis and nasal obstruction [6]. Also, features of facial pain, expectoration of tissue, epiphora, headache, vision loss, exophthalmos, and anosmia were reported with an average duration of symptoms of 3.5 months [11].

Budrukkar et al. in their study on 22 cases found 21 males and a single female, with more in younger adults. The median age was 44 years, with the youngest being a 10-year-old boy with sphenoid sinus bone erosion and positive neck nodes. All presented in advanced stages with five having intracranial extension, but neck nodes were seen only in one case. They reported varied amounts of epithelial and connective tissue elements on microscopy. Immature cartilage, chondroid cells, smooth muscle cells, bone, neuronal tissue, and squamous epithelium resembling fetal oral mucosa were reported [12]. Shorter et al. reported the first known case of this tumor with intracerebral metastasis managed via a multidisciplinary approach. They reported rare metastasis into the spinal axis, cervical lymph nodes, and respiratory tract [13].

Conclusion

Sinonasal teratocarcinosarcoma is a rare and aggressive sinonasal tumor with a hypothesized origin from somatic pluripotent stem cells of the nasal cavity. Sinonasal teratocarcinosarcoma has a proven poor prognosis with locoregional recurrence. Histopathological examination and immunohistochemistry are a must for clinching the diagnosis. Currently, no standard treatment regimen is available; surgery plus adjuvant radiotherapy with or without chemotherapy is the most common treatment modality. Early detection and surgery combined with adjuvant treatments will improve patient prognosis.

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